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## **The association between the social and communication elements of autism, and repetitive/restrictive behaviours and activities: A review of the literature.**

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## **Abstract**

Research continues to try and pinpoint the etiological role of particular genes and brain structure in autistic spectrum disorder (ASD), but despite a host of biological, genetic and neuropsychological research, the symptom profile of Pervasive Developmental Disorders (PDD) are not yet linked to etiological theory. Debate continues around whether or not there is one single dimension that incorporates the three criteria domains of social difficulties, communication deficits and repetitive or restrictive interests and behaviours as a unitary 'ASD' concept, or whether PDD as they are currently described represent the co-occurrence of separate sub-domains of developmental difficulties. Although the three criteria need to be met for a diagnosis of PDD to be made, the association between them remains unclear. This review highlights that the majority of the literature that looks at the triad of impairments suggests the symptom structure does not match that proposed by diagnostic manuals, and that the triad may no longer fit as the best way to conceptualise ASD.

## **1. Introduction**

Autism was first officially described in the Diagnostic and Statistical Manual of Mental Disorders - 3<sup>rd</sup> Edition (DSM-III; American Psychiatric Association [APA], 1980). Before this, controversy surrounded the validity of autism spectrum disorder (ASD) as a diagnostic concept, with ongoing debate as to whether or not it was best conceptualised as the earliest onset of schizophrenia (Volkmar, Bregman, Cohen, & Cicchetti, 1988). In DSM-III, infantile autism was included in the new diagnostic class of Pervasive Developmental Disorder (PDD). Diagnostic criteria for this class of disorder were based on Kanner's original description of two core features of infantile autism ('extreme aloneness' and 'preoccupation with the preservation of sameness'; Kanner, 1943) and Rutter's subsequent reappraisal of a triad of impairments (Rutter, 1968). A child had to exhibit an early disturbance with onset before 30 months, characterised by a pervasive lack of social relationships and deficits in language and/or communication, with an absence of delusions and hallucinations as found in schizophrenia (APA, 1980).

DSM-III criteria for PDD were revised for the DSM-III-R (APA, 1987). It was felt the criteria needed a more developmental focus to reflect that individuals did not stop exhibiting the disorder after early childhood, but continued experiencing difficulties throughout development (Volkmar et al., 1988). Three overarching categories described the criteria that had to be met for a diagnosis in the DSM-III-R: social dysfunction; qualitative impairments in verbal and nonverbal communication and imaginative activity, and; a restricted range of activities or interests (APA, 1987). Today, PDD continue to be characterized in DSM-IV-TR (APA, 2000) and the 10<sup>th</sup> edition of the International Classification of Diseases and Related Health Problems (ICD-10; World Health Organisation [WHO], 1992) by impairments in the three

domains of social interaction, communication, and repetitive, stereotyped behaviours and activities. Although not due for publication until 2013, a proposed revision of autistic disorder for DSM-V (APA, 1994) has merged three domains into two: social/communication deficits and fixated interests and repetitive behaviours (APA, 2010).

As such, autism is still a behaviourally defined disorder. Research continues to try and pinpoint the etiological role of particular genes and brain structure, but it is not yet linked to the symptom profile of PDD (Mandy & Skuse, 2008). Autism was traditionally conceptualised as a discrete category, qualitatively different from other presentations, but a consensus is emerging that autism is in fact a dimensional disorder reflecting difficulties at the extreme end of a continuum (Mandy & Skuse, 2008). However, debate continues around whether or not there is one single dimension that incorporates the three domains of social difficulties, communication deficits and repetitive or restrictive interests and behaviours as a unitary 'ASD' concept, or whether PDDs as they are currently described represent the co-occurrence of separate sub-domains of developmental difficulties. Although the three criteria need to be met for a diagnosis of PDD to be made, the association between them remains unclear.

Delineating the construct of autism into more than a single 'ASD' dimension could further studies of the genetic & neurobiological bases of PDD (Cuccaro et al., 2003). One method authors have used to explore the structure of autism is by using factor analysis. Factor analytic techniques are used to pull out underlying structures (known as factors or components) by identifying which items co-vary (Kline, 1994). As such, factor analysis can examine whether or not the social, communication and repetitive interests/behaviour domains of autism co-vary. If they do, they should not

show up as separate factors. However, although factor analysis can test the fit of the three factor hypothesis, some difficulties do exist. Results can be profoundly influenced by sample characteristics, size and the type of measure used, and bias can be introduced in the interpretation and the naming of particular factors (Field, 2005).

The aim of this systematic review was to try and clarify the association between the social deficits, communication impairments, and repetitive/restrictive behaviours and activities found in autism and PDD and to address the question: Does the triad of impairment still fit?

## **2. Method**

### *2.1 Search Strategy*

Key words were gathered from previous literature searches. The search terms *autis\** OR *asperger\** OR *pervasive developmental disorder* were combined with *AND struct\** and used to search Ovid databases. These were Medline (1950-May wk 1 2010), Embase (1980- wk 18 2010), Psych info (1967–May wk 2 2010), EBM Review Cochrane Database of systematic reviews (2005-March 2010), EBM Review Cochrane methodology register (2<sup>nd</sup> Quarter 2010), British Nursing Index and Archive (1985-2010). This range of databases was chosen as they cover social science and psychological research, to try to minimise database bias. The start of the search was chosen by the earliest year available on each database, in order to try to capture any possible relevant discussion pre-DSM-III (APA, 1980), when autism was first diagnostically described.

### *2.2 Inclusion and exclusion criteria*

The systematic review looked to identify papers that used DSM-IV-TR stipulated symptom dimensions (qualitative impairment in social interaction, qualitative impairments in communication and repetitive or restricted interests, behaviours and activities [RIBA]). Articles using tools that had different symptoms to those proposed by the DSM triad (e.g., arousal, affect and cognition: Eaves & Williams, 2006; social skill, communication, imagination, attention to detail and attention switching: Hoekstra, Bartels, Cath, & Boomsma, 2008) or that only contained two of domains of (e.g., only social and communication domains; Magyar & Pandolfi, 2007) were excluded. Thus, papers were excluded if they solely focused on one diagnostic criterion such as communication disorders, empathy/social cognition, or repetitive interests/behaviours or activities, rather than the triad of impairments. Similarly, studies that did not examine the diagnostic triad but focused on secondary difficulties such as challenging behaviour or specific language disorders were excluded. Studies that examined brain structure with no reference to the triad of impairments in terms of symptoms were excluded. Only papers written in English were included and two papers needed to be excluded due to sourcing difficulties (Foster, 2003; Tien, 2008; both dissertation abstracts only). One study was excluded (Soucy & Andrews, 1997) because of concern that the sample size was insufficient to justify the number of items entered into analysis (Lewis, 1995).

### *2.3 Quality Indicators*

Each study was considered by using a guideline of quality indicators devised in part from national recommendations (Scottish Intercollegiate Guidelines Network [SIGN], 2008). Each paper was considered in terms of the clarity of its research question, as well as the context of the study (does the article adequately describe the

specific circumstances under which the research was developed, carried out and completed?). The methodology used was considered, because factors such as sample characteristics, sample size, and type of measure will have a profound effect on the results obtained by factor analysis (Kline, 1994). As ASD is now generally considered to be a dimensional disorder, with autistic traits being continuously distributed throughout the general population (Mandy & Skuse, 2008), both clinical and general population samples should be able answer questions on the structure of PDD. However differences may exist across diagnosis and subtype, and therefore clarity about the population sample (including clarity about how decisions to split the sample by ability level were made) and method of diagnosis were used as additional indicators of quality. The type of analysis used was also clearly important, and given the long standing theories surrounding the structure of autism, particular weight was given to well-designed studies that used confirmatory analysis and referred to existing theory or findings.

#### *2.4 Results*

This search strategy yielded 3,922 potentially relevant citations, which was reduced to 2,538 after extraction for duplicates. All titles were examined and considered using the search criteria described above. In total 244 relevant articles were identified by title, and abstracts selected. From abstract selection, 44 studies were identified as being eligible, and full papers were sourced to confirm relevance. These papers were then examined, and 13 full papers remained.

#### *2.5 Characteristics*



The 13 papers reviewed (see table 1) represent a range of different research strategies that look to clarify the autism construct by examining the relationship found between each diagnostic criteria of the triad and the association between domains. Sample size and demographic information is highlighted, along with the diagnostic tool used in table 1.

**INSERT TABLE 1 HERE**

### **3. Review of the literature**

#### *3.1 ASD as a single dimension*

The review identified one paper that identified autism as a single dimension, and a second paper that collapsed the three domains into a single ‘autism symptom’ scale. Constantino et al. (2004) examined the factor structure of autistic traits by cluster analysis of data from all the items of the *Autism Diagnostic Interview-Revised* (*ADI-R*; Lord, Rutter, & Le Couteur, 1994) and principal factor analysis of data from the *Social Responsiveness Scale*, a third-party completed rating scale (*SRS*; Constantino, 2002). The authors explored whether the three domains of difficulties could be separated within an established clinical sample. Analysis of the *SRS* dataset revealed the presence of a primary factor that explained more than 30% of the phenotypic variance. The next three most influential factors each explained less than 7% of the variance. When the primary factor was examined, items represented all three of the DSM-IV-TR criteria of language deficits, social deficits and restricted interests or odd repetitive behaviours.

Cluster analysis of the *ADI-R* dataset yielded similar results. The authors’ analysis supported a ten factor solution, in which the first two factors were made up of

almost half of the 63 *ADI-R* items, and accounted for 27% of variation in the data. The first cluster was characterised by questions relating to social deficits, nonverbal and verbal communication deficits and sensitivity to noise. The second cluster was characterised by symptoms across all three DSM-IV-TR criteria domains, including difficulties with group play, reciprocal conversation and echolalia, and repetitive or stereotypic behaviours. However, a strong correlation between these neighbouring clusters suggested significant overlap between the two. The *ADI-R* data was then subjected to principal components analysis, in which a primary factor was found to account for 40% of the variance. This factor included items from all three criteria domains for PDD. Constantino et al. (2004) concluded that their data supported the presence of a single underlying variable of autistic spectrum conditions, manifesting characteristics across the three domains. They did not find evidence of independent sub-domains of deficits in social skills, language or repetitive/restrictive activities and behaviours.

Constantino and colleagues used exploratory factor analysis due to sample size limitations. Although helpful in exploring data, it is not optimal in testing hypotheses or answering questions on structure, the area of interest for the current review, when theories already exist (Field, 2005). Furthermore, each sample was small for the number of items within the assessment tool, and the sample had a mix of diagnoses and symptom severity. The authors argue that the inclusion of individuals with a wide range of autistic symptomatology is important to avoid amplification of a specific structure within a narrow range of severity (for instance, a structure that may only exist at the extreme end). However, they did not report whether there was a difference in structure between groups before combining their sample. Another note of caution is that the *SRS* is heavily orientated towards social language, which could have given

extra weight to the single factor finding, although this would not explain their finding using the *ADI-R*, a tool that, as the authors report, is recognised as a ‘gold-standard’ parental report.

Szatmari et al. (2002) also examined whether PDDs are composed of distinct dimensions of autistic symptoms or a single ‘autism’ construct, but they included level of functioning as a possible dimension that could account for the varying symptoms or phenotypic variation observed within PDDs. They used exploratory techniques on data from the *ADI* (1989 version: LeCouteur, Rutter, Lord, & Rios, 1989) and the *Vineland Adaptive Behaviour Scales* (*VABS*: Sparrow, Ball, & Cicchetti, 1984). Their sample was made up of two groups, one ‘lower functioning’, as measured by the *VABS* with a diagnosis of autism, and one who met their criteria for high functioning autism (HFA) or Asperger Syndrome (AS). Of note however, is that the authors decided not to use IQ as a measure of level of functioning, but relied on the *VABS* score. Measurement of IQ could have been used as an additional measure of the validity of each ‘level of functioning’ subgroup. Diagnosis of ASD was made by a best-estimate diagnostic procedure based on DSM-III-R criteria, using the opinions of two psychiatrists, before the *ADI* was completed. If no discrepancy between reports were noted, they joined the sample. As there was no algorithm from the *ADI* for AS, the authors derived their own on the basis of previous work (details of their diagnostic process can be reviewed in Szatmari et al., 2000).

Principal components analysis (PCA) was performed on the *ADI* and *VABS* ratings within each group. The authors also analysed the pooled sample after checking that the factor structure was stable across both groups. This is important because characteristics of the sample can profoundly influence the results of factor analysis. As both groups showed similar factor structure, they were able to be combined to

provide more precise estimates of factor loadings. Two factors were identified that explained almost 70% of the variance. Factor 1 represented ‘level of functioning,’ being made up of the scores on the *VABS*, whereas factor 2 comprised of ‘the autism symptom factor’, the scores from the *ADI*.

This study used exploratory techniques despite a previously proposed theoretical structure being reported. Confirmatory factor analysis may have been more appropriate, as PCA is inductive and hypothesis generating. The authors acknowledge that their population was not a representative sample of children with PDD, however, they concluded their results were indicative of two dimensions underlying the phenotypic variation in autism; a symptom domain and a level of functioning domain. They conclude that the three domains should be collapsed into one single scale of ‘autism symptom’, but that a single dimension that focuses on ASD is perhaps an oversimplification.

### *3.2 Separate sub domains corresponding to DSM-IV-TR criteria*

Other studies suggest PDDs are made up of at least three separate sub domains of difficulty that correspond directly to DSM-IV-TR criteria; social deficits, communication deficits and repetitive/restrictive behaviours and activities i.e., that there is not single ‘autism’ construct. Lecavalier and colleagues (Lecavalier, Gadow, DeVincent, Houts, & Edwards, 2009) investigated the structure of PDD symptoms by using a well designed study and confirmatory factor analysis. They used a large sample of children with the full range of symptom severity, diagnosed using DSM-IV criteria. Diagnosis was made following parent interview and observation of the child, comprehensive developmental history and educational evaluation, and both parents and teachers completed the *Early Childhood Inventory (ECI-4)*: Gadow & Sprafkin,

2000) and the *Child Symptom Inventory-4 (CSI-4: Gadow & Sprafkin, 2002)*. These are DSM-IV referenced rating scales with good psychometric properties.

Analyses focused on the entire sample and separate subgroups divided by age, diagnosis and cognitive ability. The authors tested a one factor model, a two factor model of social-communication items as one item and repetitive/restricted behaviours as the other, and a three factor model corresponding to the DSM-IV triad of impairments. Their results clearly favoured the three factor solution for both teacher and parent data, with the one and two factor models yielding poor fits for both groups of informant regardless of subject characteristics.

Dworzynski, Happe, Bolton and Ronald (2009) used exploratory factor analysis on data from the *Developmental and Wellbeing Assessment (DAWBA; Goodman, Ford, Richards, Gatward, & Meltzer, 2000)* to explore the factor structure of ASD. The *DAWBA* is an interview package used to generate DSM-IV and ICD-10 diagnoses, administered by trained lay interviewers over the telephone. The *DAWBA* is not ASD specific but is used by the Office of National Statistics, so only questions relating to ASD were administered. This diagnostic procedure does not correspond to best practice (e.g., SIGN, 2007). Exploratory factor analysis identified five factors which accounted for just over 45% of all variance. Factor one (accounting for the majority of the variance at just over 16%) was concerned with social behaviours and impaired play, with communication difficulties accounted for by factor two, and language milestones factor four. Factors three and five were similar, both covering repetitive/restrictive interests and behaviours, but with the emphasis on 'repetitiveness' and 'insistence on sameness' respectively. Thus, this data recognised the triad described by DSM-IV criteria, but split both communication difficulties and repetitive interests/behaviours and activities into two further factors. The sample was

not large enough to determine whether or not the authors' solution was a better fit than that described by Lecavalier and colleagues (2009).

### *3.3 Separate sub domains not described by DSM-IV-TR criteria*

Symptom domains were examined by van Lang et al. (2006) who used data from the *ADI-R* algorithm, rather than raw scores, to test the goodness-of-fit of five different models. The first and second model corresponded to the DSM-IV-TR triad, with the first applying to participants' behaviour at age 4-5 years, and the second to current behaviour. Their third factor model was hypothesised from the authors' earlier explorative findings with three factors; impaired social communication, stereotyped language and behaviours, and impaired make-believe and play skills. The fourth model was constructed as a single 'autistic features' symptom domain, and the fifth a two factor model consisting of 'impaired social communication' and 'stereotyped language and behaviours'.

A robust Maximum Likelihood (ML) estimation procedure was used to offset the non-normal distribution of the data, examining the goodness-of-fit of each of their suggested models. Both the DSM based models encountered difficulties as the domain of impaired social interaction and domain of impaired communication were highly correlated, and so could not be properly estimated (van Lang et al., 2006). The authors' third hypothesised three factor model fitted the data better than any of the others, although this still only explained 34% of the variance. Although this model was based on the symptomatology of autism, it had a different structure than that of the DSM-IV-TR triad. Instead, 'impaired social communication' accounted for information about difficulties in both verbal and nonverbal social communication, 'impaired make-believe and play' described the fundamental lack of play skills in

play with peers and self, and ‘stereotyped language and behaviour’, described restrictive characteristics in speech and behaviour. Thus, their model did not fit the triad.

The model described by van Lang and colleagues (2006) had been constructed from two exploratory studies that were included in the sample, so an independent cross-validation is required. In addition, although their model was stable for the sample with intellectual disability, it is unknown if it would continue to be stable in higher functioning individuals. Overall, however, this was a well designed study that led the authors to conclude that their model offers a better representation of the symptom structure of autism than the DSM-IV-TR triad.

Other authors have also indicated that the triad is not the ‘best fit’. Tadevosyan-Leyfer et al. (2003) performed exploratory principal components analysis to break up the autism phenotype into genetically relevant components. Common items from the *ADI* and *ADI-R* were used, rather than just the algorithm items. The group identified a model with 6 factors that accounted for 41% of the variance, and validated their model with a small independent sample. One of the factors they identified, ‘social intent’, is very similar to the ‘impaired social communication’ factor identified by van Lang et al. (2006), but the models differed regarding stereotyped language and behaviours. The authors concluded that their results supported a move to return to two diagnostic criteria as originally proposed by Kanner in 1943, focussing on the social deficits and the ‘insistence of sameness’. They concluded the two current standard criteria for autism, communication and social interaction, are not independent.

Georgiades and colleagues (2007) also used data from the *ADI-R* algorithm to search for the underlying structure of the autism phenotype. They used principle

components analysis then confirmatory factor analysis, in a well designed study, with a comprehensive best-estimate method for diagnosis. They had a large sample size, with all participants having been diagnosed with a range of PDDs. Results suggested that a three factor solution was the best fit, accounting for 50% of the variance. The authors described these three factors as ‘social-communication’, ‘inflexible language and behaviour’, and ‘repetitive sensory and motor behaviour’. The combined social communication factor covers both domains as described in the DSM-IV and lack of varied spontaneous social or make-believe play. The repetitive/restrictive domain is split over two factors, one covering stereotyped language and preoccupation with patterns of interest, and the other including sub-domains that measure stereotyped motor mannerisms and preoccupation with sensory stimuli and objects. They conclude that the autism symptom phenotype is made up of three domains that are different to those described by the DSM-IV, and are certainly not composed of a single autism domain.

Not all studies have concentrated on data from the *ADI-R*. Wadden, Bryson, and Rodger (1991) performed a factor analysis on the *Autism Behaviour Checklist* (*ABC*; Krug, Arick, & Almond, 1980a, 1980b) to explore the structure of this diagnostic instrument and its diagnostic discriminant ability. They concluded the *ABC* taps into three different aspects of autistic behaviour: ‘nonresponsive’ (an underlying failure to respond to the environment and social inattentiveness); ‘aloof/repetitive’ (both verbal and motor repetitiveness, poor eye contact) and; ‘infantile/aggressive’ (temper tantrums, aggression, communicating by gestures). Although they were not addressing the structure of autism per se, within their data they found no evidence for a single autism factor, and their model did not fit the traditional triad. However, their



sample was made of a clinical and control group analysed together, and there is no report of checking for a stable factor structure across both groups.

Posserud et al. (2008) investigated the factor structure of the *Autism Spectrum Screening Questionnaire (ASSQ)*: Ehlers & Gilberg, 1993). The aim of their study was to clarify how to separate ASD from cases of social impairment due to other causes. The *ASSQ* is a teacher and parent self-report questionnaire covering social interaction, verbal and nonverbal communication and restricted/repetitive behaviours and motor clumsiness. Principal components analysis and confirmatory factor analysis supported a three factor solution for both respondent groups. The first factor was labelled ‘social difficulties’, the second ‘tics/motor/OCD’ as it included many items relating to repetitive, stereotypic behaviour in autism. The third factor was labelled ‘autistic style’ denoting the cognitive style and language characteristics seen in high-functioning individuals with autism. They concluded it was this third factor that was key in identifying the qualitative difference in difficulties between autism and other causes of social impairment. Posserud et al. (2008), however, stress that their data were not intended to be interpreted as an analysis of dimensions within autism, as their sample was not a PDD clinical population, but was obtained from the general population. This sample was chosen as the *ASSQ* was originally developed as a screening measure to identify children who may need further clinical assessment, rather than as an instrument to confirm a diagnosis of ASD. However, studies with the general population are still helpful in considering the structure of autism, as autistic traits have been proposed to be continuously distributed throughout the general population (see Mandy & Skuse, 2008).

### *3.4 Merging of DSM-IV-TR criteria*

Although some studies have suggested that social and communication difficulties are separate domains, other research suggests that they load onto one single factor. Lecavalier et al. (2006) examined the algorithm items of the *ADI-R* to assess its validity. Exploratory and confirmatory factor analysis found a three factor model fit the data best, explaining 38% of the variance. However, they used the same sample to compare the results of the exploratory functional analysis with the *ADI* algorithm modelled on DSM-IV-TR criteria, despite the identified risk of capitalising on chance. While their model closely resembled the DSM-IV-TR diagnostic symptom domains, there was one discrepancy, in that all nonverbal communication items were associated with the social factor. This factor accounted for just over 21% of the variance. ‘Communication’ needed to be split between nonverbal and verbal skills, as social deficits and communication deficits did not appear to be distinct. Although Lecavalier and colleagues (2006) were assessing the validity of the *ADI-R*, their psychometric results can add to the debate about the behavioural dimensions of the autistic phenotype, in that their study highlights the overlapping nature of symptoms regarded as separate domains.

Frazier, Youngstrom, Kubu, Sinclair, and Rezai (2008) also examined the factor structure of the *ADI-R* algorithm using both exploratory and confirmatory factor analysis methods, but used a much larger sample, from a longitudinal research programme, than that of Lecavalier and colleagues (2006). They also examined the factor structure across two age groups. Again, their data indicated that the factor structure of the *ADI-R* used to diagnose autism is different to that described by the DSM-IV-TR triad. Instead, a two factor structure was presented, with restricted/repetitive and stereotyped behaviour loaded with stereotyped language onto one factor, and impairments in social interaction and communication combined

together on a second factor. The authors suggest autism domains may need to be restructured to more accurately reflect the strong relationship between social and communication impairments, and to separate them from stereotyped and repetitive behaviours.

Snow, Lecavalier, and Houts (2009) investigated the factor structure of the *ADI-R* by using every item rather than the algorithm, but used the same longitudinal dataset as Frazier et al. (2008) as part of their large sample. They also explored the convergence of *ADI-R* performance with measures of adaptive, language, and cognitive functioning. Based on best fit indices, a two factor model solution was presented, consisting of social/communication items and restricted/repetitive behaviour items. This model was a better fit than the traditional three-domain model based on diagnostic criteria or a single ‘autism’ factor solution.

The dimensional structure of the autism phenotype was also investigated by Kamp-Becker, Ghahreman, Smidt, and Remschmidt (2009) in a population of high functioning individuals who attended their clinic for assessment. Two exploratory factor analyses were conducted, one on ‘early development’ data from the *ADI-R* and one on ‘current presentation’ data from the *Autism Diagnostic Observation Scale (ADOS-G; Lord et al., 2000)*. Factor analysis on the *ADI-R* supported a four factor solution. The first factor was named ‘social communication’ and explained approximately 17% of the variance, and combined items from the original social interaction and communication domains. The second factor was named ‘anxiety and compulsions’ and included circumscribed interests and verbal rituals. The third factor was characterised by ‘stereotyped behaviour’, both verbal and nonverbal. The final factor was described as ‘inadequate behaviours’. Comparison of the autism and non-

autism groups showed considerable difference between the ‘social communication factor’ and the ‘anxiety and compulsions factor’.

Kamp-Becker et al. (2009) selected a five factor solution for factor analysis of the *ADOS-G* dataset, which accounted for 57% of the variance. The first factor covered ‘social communication’ items within a single sub-domain, explaining 26% of the variance. The second factor was named ‘non/verbal behaviour’ and included items influenced by eye contact and speech abnormalities. The third factor was named ‘hyperactivity’, fourth ‘stereotyped behaviour’ and fifth ‘interests and compulsions’. Again, comparisons of the autism and non-autism group showed significant differences for the social communication factor as well as the non/verbal factor. They concluded that the AS/HFA phenotype was structured by dimensions that differed to the conceptualisation of DSM-IV-TR criteria, particularly because the social interaction and communication domains were so closely related that they emerged as a single factor.

#### **4. Discussion**

The review highlighted a general lack of consistency about the number and structure of factors identified, and no definite agreement on the association between the social and communication elements of autism, and repetitive/restrictive behaviours and activities was found.

The majority of studies reviewed used exploratory factor analysis (including principle components analysis) rather than confirmatory factor analysis to investigate the structure of autism. As confirmatory factor analysis looks to assess the fit of a proposed model to see how well it captures the covariance between each item, it may be more suited to answering questions on the structure of PDD symptoms (Field,

2005). However, confirmatory techniques require a larger sample size and so most reviewed investigations used principal components analysis or exploratory factor analysis to explore the data. As discussed, factor analysis is sensitive to sample size, and even slight variations in sample composition and factor extraction criteria may give different results (Kline, 1994). The wide range of different sample sizes in the reviewed studies may explain some of the variation in conclusions between them.

There are also a number of alternative explanations for these differences. Interpretation and the naming of particular factors are dependent on the author's understanding of the data, which introduces a subjective element. In addition, although nine of the identified studies examined data from the *ADI* or *ADI-R*, some authors only used items from the diagnostic algorithm (e.g., Lecavalier et al., 2006), others used all sub-domain scores (e.g., Georgiades et al., 2007) or a selection of items from the *ADI* and *ADI-R* (e.g., Kamp-Becker et al., 2009; Tadevosyan-Leyfer et al., 2003).

No two studies had the same design, with different samples, age ranges and diagnoses, as well as different diagnostic tools being used. Populations included in the analyses differed, with some studies including a broad range of autistic symptoms (e.g., Constantino et al., 2004) and others focusing on a narrow range of autistic traits, which could artificially inflate the association between symptoms and dimensions (Mandy & Skuse, 2008). Similarly, not all studies used data from the same questionnaire. As some questionnaires are designed as screening instruments to identify individuals with traits in need of further assessment, and others aim for definitive diagnosis, different questionnaires emphasise different core features. Also, different questionnaires were used across populations of varying cognitive ability.

This is all likely to go some way to explaining the difference in factor structure proposed by the reviewed papers.

The studies by Szatmari et al. (2002) and Constantino et al. (2004) differed from the other reviewed papers, in that their results combined the diagnostic criteria into a single 'autism' factor. However, Constantino and colleagues acknowledge that their sample was not large enough to reliably fit the goodness of fit of their single factor model and Szatmari et al note that they did not use a representative sample of children with ASD. They also found differences between their two groups that appeared to relate to level of functioning, suggesting further differences in construct. It may also be that the array of behaviours that are displayed by individuals with ASD and the different ways in which these are manifested presents a complexity that is difficult to capture in a single 'autism' factor.

The study by Wadden et al. (1991) resulted in factor structures that did not correspond to DSM-IV-TR criteria. However, Wadden and colleagues used ASD and non-ASD groups, meaning that factors within the clinical data may have been missed, as between-groups variance on items that discriminate ASD from non-ASD can mask differential item variance. Only two studies, both by the same group, identified three factors that corresponded directly to DSM-IV-TR criteria of social deficits, communication deficits and repetitive behaviour (Lecavalier et al., 2006; Lecavalier et al., 2009). However, the former study was examining the psychometric properties of the *ADI-R* rather than behavioural dimensions, and the latter did report a strong correlation between social and communication scores. Similarly, although Dworzynski et al. (2009) identified three criteria described by DSM-IV-TR, they found that social behaviours and communication were correlated. ASD phenotypic correlations were highest between social and communication impairments, and

weakest between communication/language and repetitive/restrictive behaviour scores. Similarly, multiple regression showed that the best predictive relationship was between social and communication items, whereas repetitive/restrictive interest scores were not significantly predicted by any other domain. As one of the key functions of communication is social and that there is a bi-directional relationship between social interaction and communication it is perhaps unsurprising that a correlation was found between the two.

The majority of analyses resulted in authors recommending a move towards conceptualising social deficits and communication deficits as being a shared social-communication factor, with repetitive/restrictive behaviours and activities being a separate domain (Frazier et al., 2008; Georgiades et al., 2007; Kamp-Becker et al., 2009; Snow et al., 2009; van Lang et al., 2006). This would be in line with proposed DSM-V amendments. Despite finding that a six factor model suited their data best, Tadevosyan-Leyfer et al. (2003) also concluded their data suggested a move towards a two factor model of social communication deficits and repetitive interests and behaviours. These studies give weight to the suggestion that social and communication symptoms should be combined conceptually into one core domain of impairment, distinct from the restricted/repetitive behaviour domain.

#### *4.1 Implications*

Understanding the structure of autism symptoms can improve diagnostic and classification systems, as it is possible that the three-domain conceptualisation of autism does not correctly describe the disorder. This could in theory contribute to unreliable diagnoses. By empirically examining the structure of autism symptoms, we can refine diagnostic procedures, as well as consider different phenotypes. The

studies within this review examined the structure of ASD and were chosen as they included all three diagnostic criteria. Even within this sample however, the literature raises questions about differences in symptomatology in low and higher functioning individuals with ASD, and whether the separate symptom domains have different developmental trajectories.

Research has suggested that the severity of repetitive interest/behaviours is inversely correlated to IQ (Cuccaro et al., 2003, Szatmari et al., 2006) and Georgiades et al. (2007) found children with AS had high scores on the inflexible language and behaviour factor but low scores on the repetitive sensory and motor behaviour factor. Kamp-Becker et al. (2009) also found a significant correlation between their factor ‘stereotyped behaviour’ and performance IQ. It may be that there is a weaker relationship between social-communication symptoms and repetitive/restrictive behaviours and activities in high functioning people with ASD. Interestingly, Lecavalier et al. (2009) reported that different subgroups included in their analyses impacted the fit of their model. Samples of children with AS fit the DSM-IV-TR three factor model best, whereas data from children with a diagnosis of autism did not fit the model so well.

Future studies could continue to use factor analysis to examine the ‘fit’ of the autistic triad in low and higher functioning individuals with ASD, and consider whether the separate symptom domains have different developmental trajectories. However, despite three decades of exploration there is still no clear answer about the triad’s empirical relevance. It may be that a wider exploration of areas of human development is required to capture all possible domains of impairment in individuals with ASD.



## **5. Conclusion**

This review has suggested that although there are alternative ways to understand the structure of autism, the majority of the literature that looks at the triad of impairments suggests the symptom structure does not match that proposed by the DSM-IV-TR, and that the triad may no longer fit as the best way to conceptualise ASD. Instead, social and communication deficits show an association that suggests they should be considered together as a single domain, and repetitive/restrictive behaviours and activities considered as a separate symptom domain. Refining the structure of the autistic phenotype can provide valuable information for both diagnostic procedures and genetic research, as the identification of core symptoms might be useful in genetic linkage studies.

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Table 1: Summary of reviewed papers

| Author                              | Sample size | Dx   | Age         | Diagnostic Tool   | Statistical technique used                | Number of factors  |
|-------------------------------------|-------------|--|-------------|---|---|--|
| Wadden ,<br>Bryson &<br>Rodger 1991 | 123         | ASD or<br>Intellectual<br>Disability (ID)                    | 6-<br>15yrs | ABC, mental age and<br>chronological age                                | Exploratory<br>factor analysis<br>(EFA)   | 3 factors:<br>1. Non-responsive<br>2. Aloof/Repetitive<br>3. Infantile/Aggressive  |
| Szatmari et al.<br>2002             | 129         | PDD (no ID)  | 4-<br>17yrs | ADI (1989 version) and VABs   | Principal<br>components<br>analysis (PCA) | 2 factors:<br>1. Autistic symptoms<br>2. Level of functioning  |
| Tadevosyan et<br>al. 2003           | 292         | ASD  | 2-<br>47yrs | ADI and ADI-R (common items)  | PCA                                       | 6 factors:<br>1. Spoken language<br>2. Social intent<br>3. Compulsions<br>4. Developmental<br>milestones<br>5. Savant skills<br>6. Sensory aversions |
| Constantino et<br>al. 2004          | 226         | PDD &<br>unspecified<br>developmental<br>disorder (no<br>ID) | 4-18<br>yrs | ADI-R (all 12 subdomain scores)<br>Social Responsiveness Scale<br>(SRS) | Cluster analysis<br><br>PCA               | Single underlying 'autism'<br>factor   |
| Lecavalier et<br>al. 2006           | 226         | PDD or<br>ADHD   | 5-<br>11yrs | ADI-R (algorithm)   | EFA                                       | 3 factors:<br>1. Social<br>2. Communication<br>3. Repetitive Behaviour   |

|                        |      |                            |           |  |                                    |  |
|------------------------|------|----------------------------|-----------|--|------------------------------------|--|
| Van Lang et al. 2006   | 255  | ASD & typically developing | 4-20yrs   | ADI-R (algorithm)                            | Confirmatory factor analysis (CFA) | 3 factors:<br>1. Impaired social communication<br>2. Impaired make believe and play<br>3. Stereotyped language and behaviour |
| Georgiades et al. 2007 | 209  | PDD                        | 2-40yrs   | ADI-R (all subdomain scores)                 | PCA & CFA                          | 3 factors:<br>1. Social-Communication<br>2. Inflexible Language and Behaviour<br>3. Repetitive sensory and motor behaviour   |
| Posserud et al. 2008   | 6229 | General population only    | 7-9yrs    | ASSQ   | PCA and EFA                        | 3 factors:<br>1. Social function<br>2. Autism-associated problems<br>3. Cognitive style associated with HFA/AS               |
| Frazier et al. 2008    | 1170 | ASD                        | 2-46 yrs  | ADI-R (algorithm)                            | PCA & CFA                          | 2 factors:<br>1. Stereotyped language & RIBA<br>2. Impairments in social interaction & communication                         |
| Dworzynski et al. 2009 | 189  | PDD                        | 10-12 yrs | Development and Wellbeing Assessment (DAWBA) | PCA                                | 5 factors:<br>1. Social Behaviours<br>2. Communication<br>3. Language delay  |

|                         |      |     |         |  |             |   |
|-------------------------|------|-----|---------|--|-------------|---|
|                         |      |     |         |  |             | <ul style="list-style-type: none"> <li>4. RIBAs (repetitive)</li> <li>5. RIBAs (insistence of sameness)</li> </ul>  |
| Kamp-Becker et al. 2009 | 140  | PDD | 6-24yrs | ADI-R (items)<br>ADOS-G (module 3 and 4) | EFA         | <p>ADI-R 4 factors:</p> <ul style="list-style-type: none"> <li>1. Social Communication</li> <li>2. Anxiety and Compulsions</li> <li>3. Stereotyped Behaviour (verbal and nonverbal)</li> <li>4. Inadequate Behaviours</li> </ul> <p>ADOS-G 5 factors</p> <ul style="list-style-type: none"> <li>1. Social communication</li> <li>2. Non/Verbal Behaviour</li> <li>3. Hyperactivity</li> <li>4. Stereotyped Behaviour</li> <li>5. Interests and Compulsions</li> </ul> |
| Lecavalier et al. 2009  | 730  | PDD | 3-12yrs | ECI-4 & CSI-4 (DSM-IV reference scales)  | CFA         | <p>3 factors:</p> <ul style="list-style-type: none"> <li>1. Social</li> <li>2. Communication</li> <li>3. Repetitive Restricted Behaviours</li> </ul>  |
| Snow et al. 2009        | 1961 | PDD | 4-18yrs | ADI-R (algorithms only)                  | EFA and CFA | <p>2 factors:</p> <ul style="list-style-type: none"> <li>1. Social/communication items</li> <li>2. Restricted/repetitive behaviour items</li> </ul>   |